Journal of the Saudi Society of Dermatology & Dermatologic Surgery (2013) 17, 21–23

King Saud University

Journal of the Saudi Society of Dermatology & Dermatologic Surgery

www.ksu.edu.sa
www.jssdds.org
www.sciencedirect.com

CASE REPORT

Lichen planus pigmentosus in a 3 year old Kashmiri boy: Rare presentation

Iffat Hassan *, Parvaiz Anwar Rather, Gousia Sheikh, Saima Aleem

Department of Dermatology, STD and Leprosy, Govt. Medical College, Srinagar, J&K, India

Received 6 August 2012; revised 16 October 2012; accepted 23 October 2012
Available online 24 November 2012

KEYWORDS

Lichen planus;
Lichen planus pigmentosus;
Childhood lichen planus pigmentosus

Abstract Lichen planus (LP) is a chronic inflammatory dermatological condition usually affecting adults, but rare in children. Lichen planus pigmentosus (LPP) is an uncommon variant of LP seen mostly in India or Middle East. LPP as such is rare and its presentation as childhood LPP rarer. We report a case of LPP in a 3 year old Kashmiri male child, which is a rare presentation.

1. Introduction

Lichen planus (LP) is a chronic inflammatory dermatological condition usually affecting adults, but rare in children (Kanwar and De, 2010). Lichen planus pigmentosus (LPP), an uncommon variant of LP seen mostly in India or Middle East, is characterized by diffuse, mottled, reticulated or perifollicular hyperpigmented, dark brown macules differing from classical LP by longer clinical course without pruritus or the scalp, nail, or mucosal involvement (Bhutani et al., 1974). Individual case reports for the childhood LPP are rarely found in literature. Keeping this in view, we report the rare case of childhood LPP.

2. Case report

A 3 year old male child, outcome of a consanguineous marriage, borne by normal delivery, first in birth order, presented to our out-patient department in June 2012, with 3 months duration of asymptomatic hyper pigmented skin lesions on the forehead, the neck, the trunk and the upper extremities, which started from the trunk. One month prior to this, he suffered from an episode of varicella. There was no history of preceding trauma, significant drug or family history. The child had normal physical and mental development and no delayed milestones. General physical and systemic examinations were normal. On cutaneous examination, there were bilaterally symmetrically distributed bluish-brown discrete and coalesced, non scaly, non atrophic, macular areas of variable size over the forehead, the neck, the trunk and the upper limbs, more predominant on proximal and flexor aspects, with almost complete sparing of extensor aspect (Fig. 1a–c). The lesions were not associated with erythema. Darrier sign was negative. There was sparing of the hands, the feet, and the lower limbs. The mucous membrane, the nail, the teeth and the hair examination were normal. Our differential diagnosis included cutaneous mastocytosis, post inflammatory hyperpigmentation, idiopathic eruptive macular pigmentation (IEMP), ashy dermatosis and lichen planus pigmentosus (LPP). Complete blood count, liver function test, kidney function test, and urine examination were normal. Punch biopsy from the lesion on histopathologic examination, under haematoxylin and eosin staining and special stains for mast cells, showed mild ortho-

* Corresponding author. Tel.: +91 9419077667.
E-mail address: hassaniffat@gmail.com (I. Hassan).

Peer review under responsibility of King Saud University.

ELSEVIER Production and hosting by Elsevier

2210-836X © 2012 King Saud University. Production and hosting by Elsevier B.V. All rights reserved.
http://dx.doi.org/10.1016/j.jssdds.2012.10.001

Figure 1 BL symmetrical bluish brown macules on the neck, the proximal limbs, front of the trunk (A and B) and back of the trunk (C).

keratosis, focal basal cell vacuolation, mild chronic lymphocytic infiltrate in the subepithelium, prominent pigment incontinence, negative toluidine blue stain (Fig. 2), thereby excluding mastocytosis, IEMP and post inflammatory hyper pigmentation. Ashy dermatosis and LPP appear histopathologically similar. Ashy dermatosis was less likely because of the absence of erythema. As a case of lichen planus pigmentosus, the patient was put on griseofulvin and is being reported for its rarity.

3. Discussion

Lichen planus is a chronic inflammatory dermatological condition usually affecting adults, but rare in children (Kanwar and De, 2010). Most of the childhood LP cases have been reported from India (Kanwar et al., 1991; Kumar et al., 1993; Sharma and Maheshwari, 1999; Handa and Sahoo, 2002), with a fewer reports from other parts of the world like United Kingdom (Milligan and Graham-Brown, 1990), Italy (Cottoni et al., 1993), France (Rybojad et al., 1998), Kuwait (Nanda et al., 2001) and Mexico (Luis-Montoya et al., 2005). This observation suggests a genetic and/or environmental predisposition to LP in patients of Indian nationality.

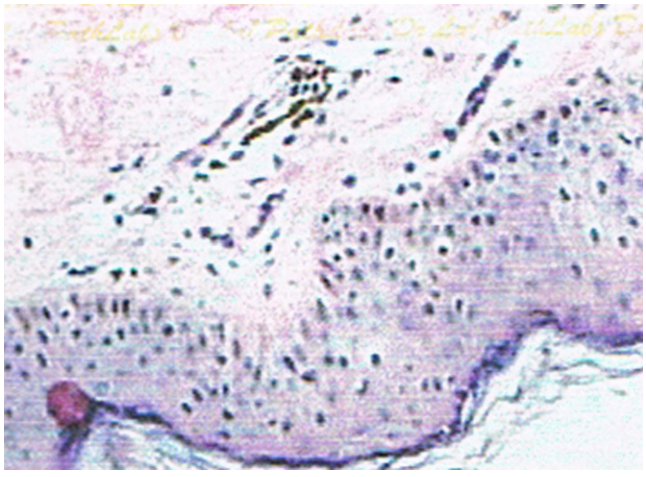

Figure 2 Photomicrograph showing orthokeratosis, focal basal vacuolation, chronic lymphocytic infiltrate in the dermis and pigment incontinence (H & E; X40).

Lichen planus pigmentosus (LPP), an uncommon variant of LP seen mostly in India or Middle East, is characterized by diffuse, mottled, reticulated or perifollicular hyperpigmented, dark brown macules differing from classical LP by longer clinical course without pruritus or the scalp, nail, or mucosal involvement (Bhutani et al., 1974). The clinical association of this entity with lesions of classical LP in about a third of patients and the demonstration of colloid bodies on histopathology prompted Bhutani et al. (1974) to consider LPP a macular variant of LP and very similar to EDP (Berger et al., 1989). The lesions lack the erythematous border of EDP.

LPP occurs most commonly on sun exposed areas such as the face, the neck, and the flexural folds (Kanwar et al., 2003), although other patterns like intertriginous, zosteriform, linear and that of non sun-exposed areas such as the thigh have been described. The cause of LPP is unknown, but an immunologic mechanism mediates its development, as well as that of lichen planus.

We did not find individual case reports for the childhood LPP in the literature, although there are few studies where some of the patients of childhood LP presented with LPP (Luis-Montoya et al., 2005; Walton et al., 2010). LPP as such is rare and its presentation in childhood rarer. In this context, we report this case of childhood LPP, the first of its kind from our state.

References

Berger, R.S., Hayes, T.J., Dixon, S.l., 1989. Erythema dyschromicum perstan and lichen planus: are they related? J. Am. Acad. Dermatol. 21, 438–442.

Bhutani, L.K., Bedi, T.R., Pandhi, R.K., Nayak, N.C., 1974. Lichen planus pigmentosus. Dermatologica 149, 43–50.

Cottoni, F., Ena, P., Tedde, G., et al., 1993. Lichen planus in children: a case report. Pediatr. Dermatol. 10, 132–135.

Handa, S., Sahoo, B., 2002. Childhood lichen planus: a study of 87 cases. Int. J. Dermatol. 41, 423–427.

Kanwar, A.J., De, D., 2010. Lichen planus in children. Indian J. Dermatol. Venereol. Leprol. 76, 366–372.

Kanwar, A.J., Handa, S., Ghosh, S., et al., 1991. Lichen planus in childhood: a report of 17 patients. Pediatr. Dermatol. 8, 288–291.

Kanwar, A.J., Dogra, S., Handa, S., Parsad, D., Radotra, B.D., 2003. A study of 124 Indian patients with lichen planus pigmentosus. Clin. Exp. Dermatol. 28, 481–485.

Kumar, V., Garg, B.R., Baruah, M.C., et al., 1993. Childhood lichen planus. J. Dermatol., 175–177.

Luis-Montoya, P., Dominguez-Soto, L., Vega-Memije, E., 2005. Lichen planus in 24 children with review of the literature. Pediatr. Dermatol. 22, 295–298.

Milligan, A., Graham-Brown, R.A.C., 1990. Lichen planus in children – a review of six cases. Clin. Exp. Dermatol. 15, 340–342.

Nanda, A., Al-Ajmi, H.S., Al-Sabah, H., et al., 2001. Childhood lichen planus: a report of 23 cases. Pediatr. Dermatol. 18, 1–4.

Rybojad, M., Moraillon, I., Laglenne, S., et al., 1998. Lichen planus in children: 12 cases. Ann. Dermatol. Venereol. 125, 679–681.

Sharma, R., Maheshwari, V., 1999. Childhood lichen planus: a report of 50 cases. Pediatr. Dermatol. 16, 345–348.

Walton, K.E., Bowers, E.V., Drolet, B.V., Holland, K.E., 2010. Childhood lichen planus: demographics of US population. Pediatr. Dermatol. 27, 34–38.